Treatment of Ruptured Saccular Aneurysms of the Fenestrated Vertebrobasilar Junction with Balloon Remodeling Technique

A Short Case Series and Review of the Literature

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Summary

Fenestration of the intracranial arteries is a relatively common occurrence. This anatomic variation may predispose to aneurysm formation at certain sites. Treatment of such aneurysms is difficult as it may occlude one of the limbs of fenestration with resultant deficit. Thus, preservation of both the limbs with adequate exclusion of the aneurysm from the circulation should be the aim of any treatment. We describe a series of four cases of ruptured aneurysms arising from a fenestrated vertebrobasilar junction treated with endovascular balloon remodeling technique.

Introduction

Fenestration of the intracranial arteries is not very rare with a reported frequency of approximately 23% in the anterior circulation and 7% in the posterior circulation [1].

Basilar artery fenestration follows vertebral artery fenestration in frequency and is reported in 0.6% of angiograms [2] and in about 5% of autopsy series [3]. Aneurysms arising from the posterior circulation are estimated to be around 15% of all intracranial aneurysms while the incidence of vertebrobasilar junction (VBJ) aneurysm is 0.5% among all intracranial aneurysms [4].

Basilar artery fenestrations are accompanied by aneurysms in 7% of cases. The incidence of fenestration in the presence of a vertebrobasilar junction aneurysm is relatively high at approximately 35% [5]. Due to the complex geometry of the fenestration, the close proximity to the brainstem and lower cranial nerves and the multiple small perforating arteries arising from the basilar artery [6], appropriate treatment, especially by open surgery of associated aneurysms, is fraught with a high risk of morbidity and mortality. Endovascular management has emerged as the first line therapy in these aneurysms with reasonably good results [4]. Aneurysms associated with fenestration require extra care due to their complex anatomy and high risk of occlusion of the dominant limb of fenestration feeding the posterior circulation. A well-planned and precise approach towards these lesions is often rewarding. We describe a short series of VBJ aneurysms successfully embolized using endovascular techniques and devices. This is followed by a systematic review of all reports in medical literature which have dealt with fenestrated VBJ aneurysms by endovascular means.

Case Series

Case 1

A 48-year-old woman presented in the emergency room with subarachnoid hemorrhage. She was a known hypertensive for the last two

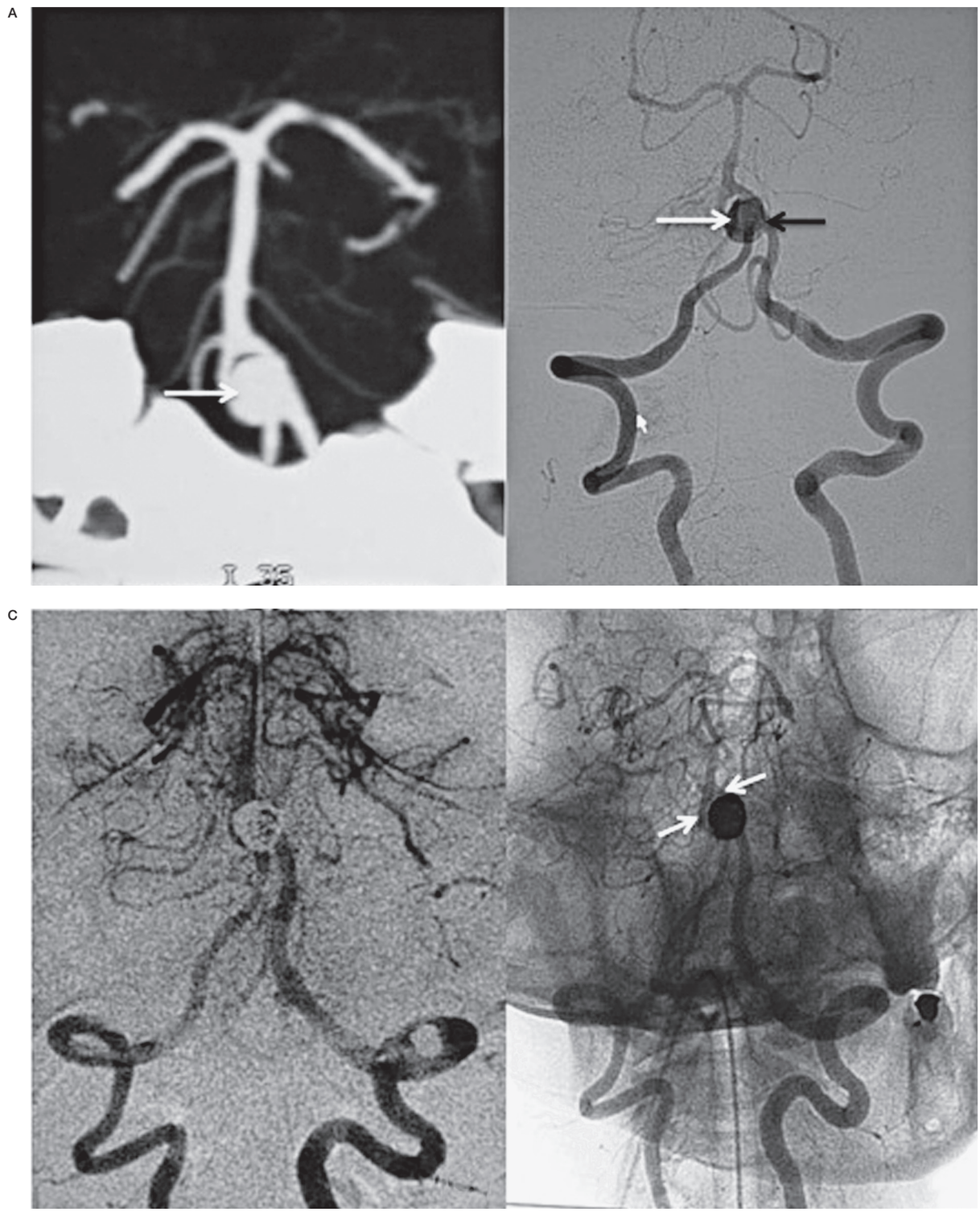

Figure 1 A) Pre procedure CT angiography. B) Digital subtraction angiography. C) Post procedure digital subtraction angiography. D) Post procedure unsubtracted angiography (D). A saccular aneurysm can be seen arising from the proximal bifurcation of the fenestration (white arrow in A and B) with incorporation of the left limb of the fenestration (black arrow in B). Post procedure images (C,D) show complete obliteration with the platinum coils with preservation of both limbs of the fenestration (arrows in D).

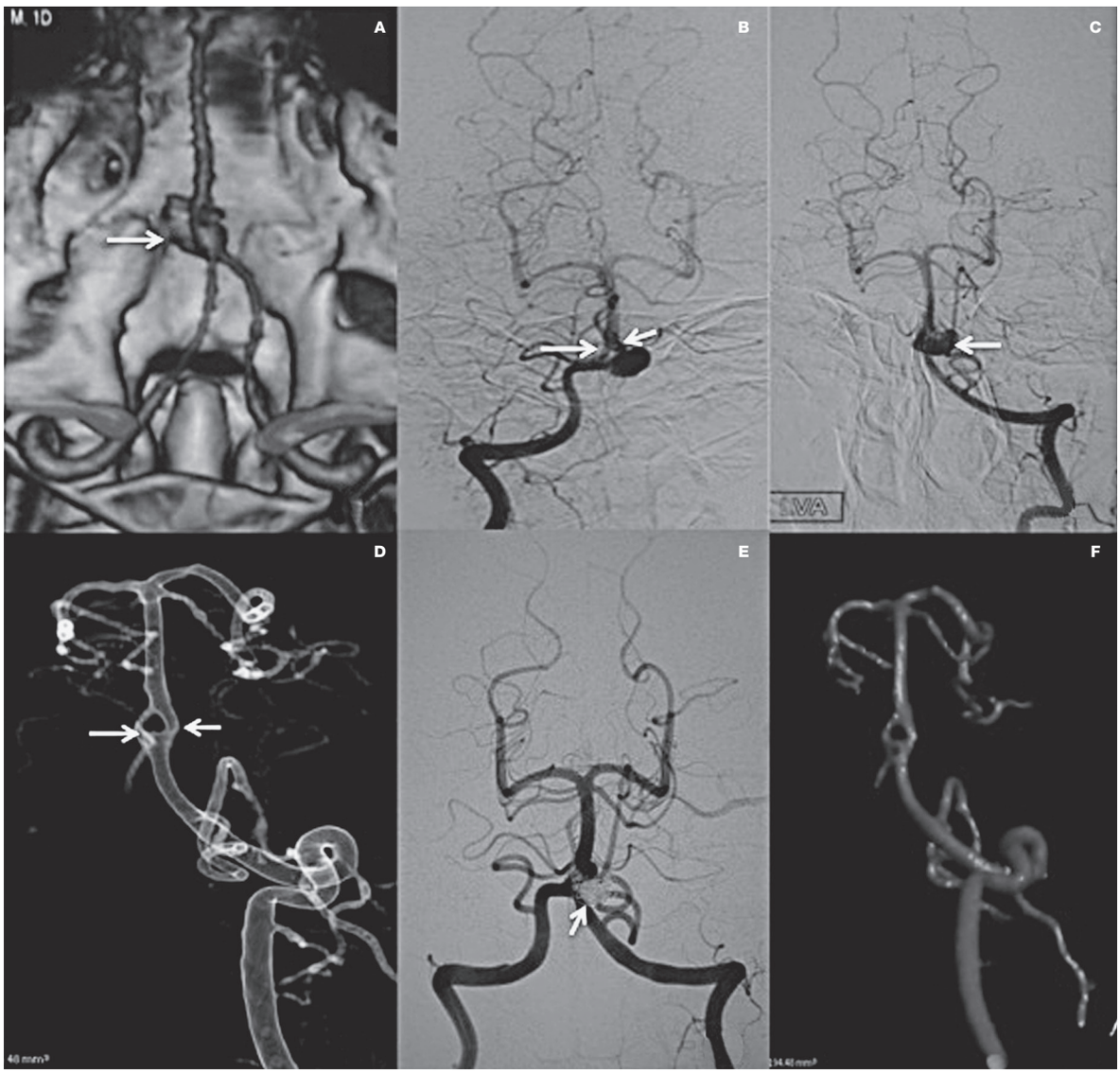

Figure 2 A) Pre procedure CT angiography. B,C) Digital subtraction angiogram of the right (B) and left (C) vertebral artery. D,F) Post procedure 3D rotational angiography. E) Post procedure digital subtraction angiography. A saccular aneurysm can be seen arising from the proximal bifurcation of the fenestration (arrow in A and C). Both limbs of the fenestration are clearly visible (arrows in B). Post procedure images (D,E) show complete obliteration of the aneurysm (arrow in E) with preservation of both limbs (arrows in D).

years on irregular medical treatment. On examination, she was conscious and oriented in time, place and person.

Non-contrast computed tomography (NC-CT) of the brain revealed posterior fossa subarachnoid hemorrhage (SAH), most marked in the prepontine cistern with intraventricular hemorrhage (Fisher grade 4). A subsequent CT angiography (CTA) showed vertebrobasilar junction (VBJ) fenestration along with a 9×8×8 mm sized saccular aneurysm arising from the bifurcation of the fenestration with partial incorporation of the left-sided limb and was directed anteriorly (Figure 1A).

Diagnostic digital subtraction angiography (DSA) confirmed the aneurysm morphology and demonstrated its immediate surrounding milieu (Figure 1B).

There was significant vasospasm in the mid basilar artery.

Endovascular coiling was performed through the left vertebral route after securing access by

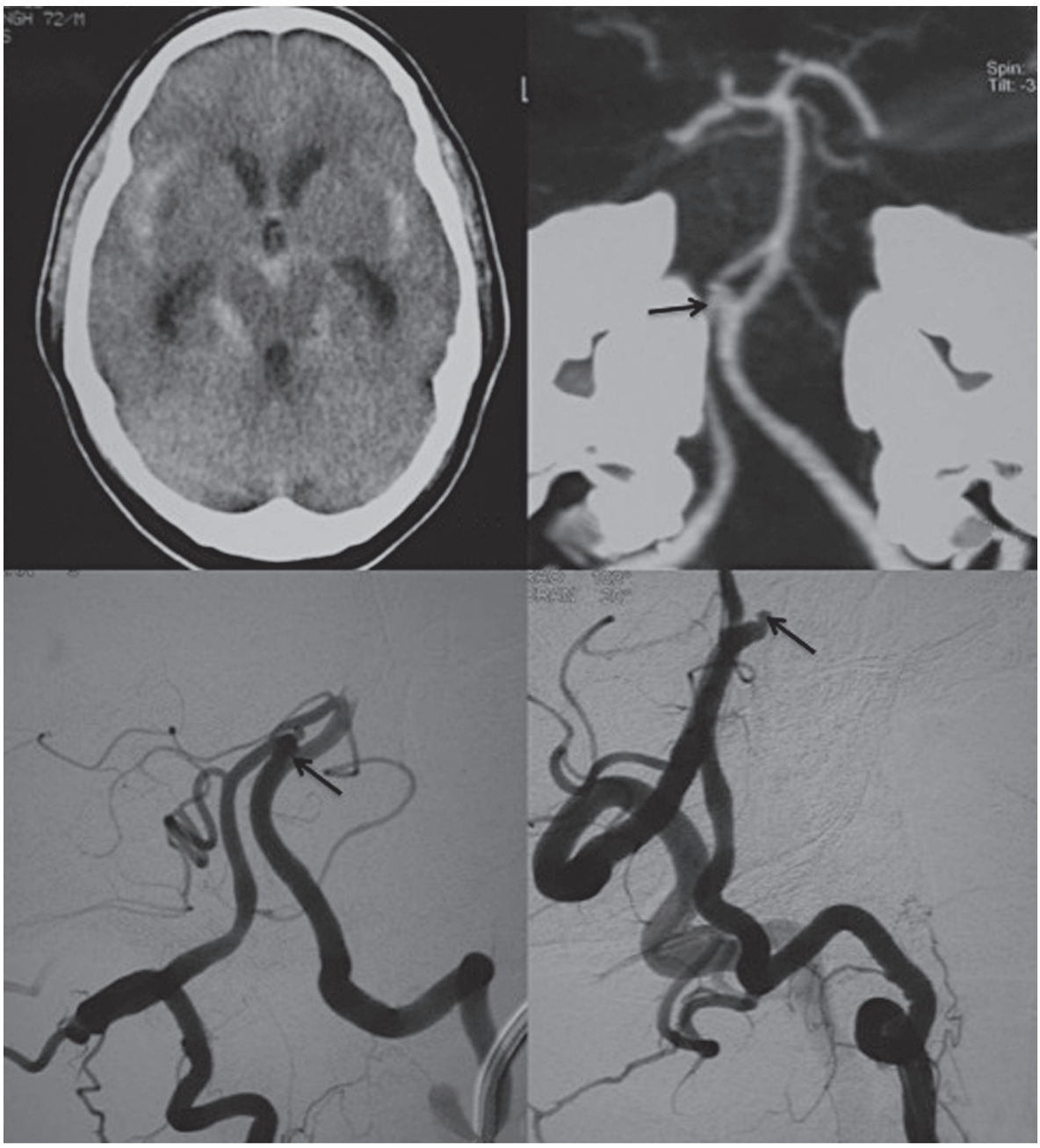

Figure 3 A-D) Patient with left subclavian proximal occlusion and subclavian steal. NCCT (A) showing diffuse subarachnoid hemorrhage with CT angiography image (B) and right vertebral AP (C) and oblique (D) angiogram images showing a saccular aneurysm arising from the proximal bifurcation of the fenestration (arrow in B) with partial incorporation of the left limb (arrow in C). Note a small daughter aneurysm arising from the dome of the aneurysm suggestive of a bleeding point (arrow in D).

placing a 6F Envoy guiding catheter (Cordis Endovascular, Miami Lakes, FL, USA) in the distal V2 segment. A total of six bare platinum Axium coils (EV3 Neurovascular, Inc, Irvine, CA, USA) were deployed in the aneurysm sac using an Echelon 10 (EV3 Neurovascular, Inc, Irvine, CA, USA) microcatheter with the aid of a Hyperglide balloon (EV3 Neurovascular, Inc, Irvine, CA, USA).

Complete aneurysm occlusion was achieved

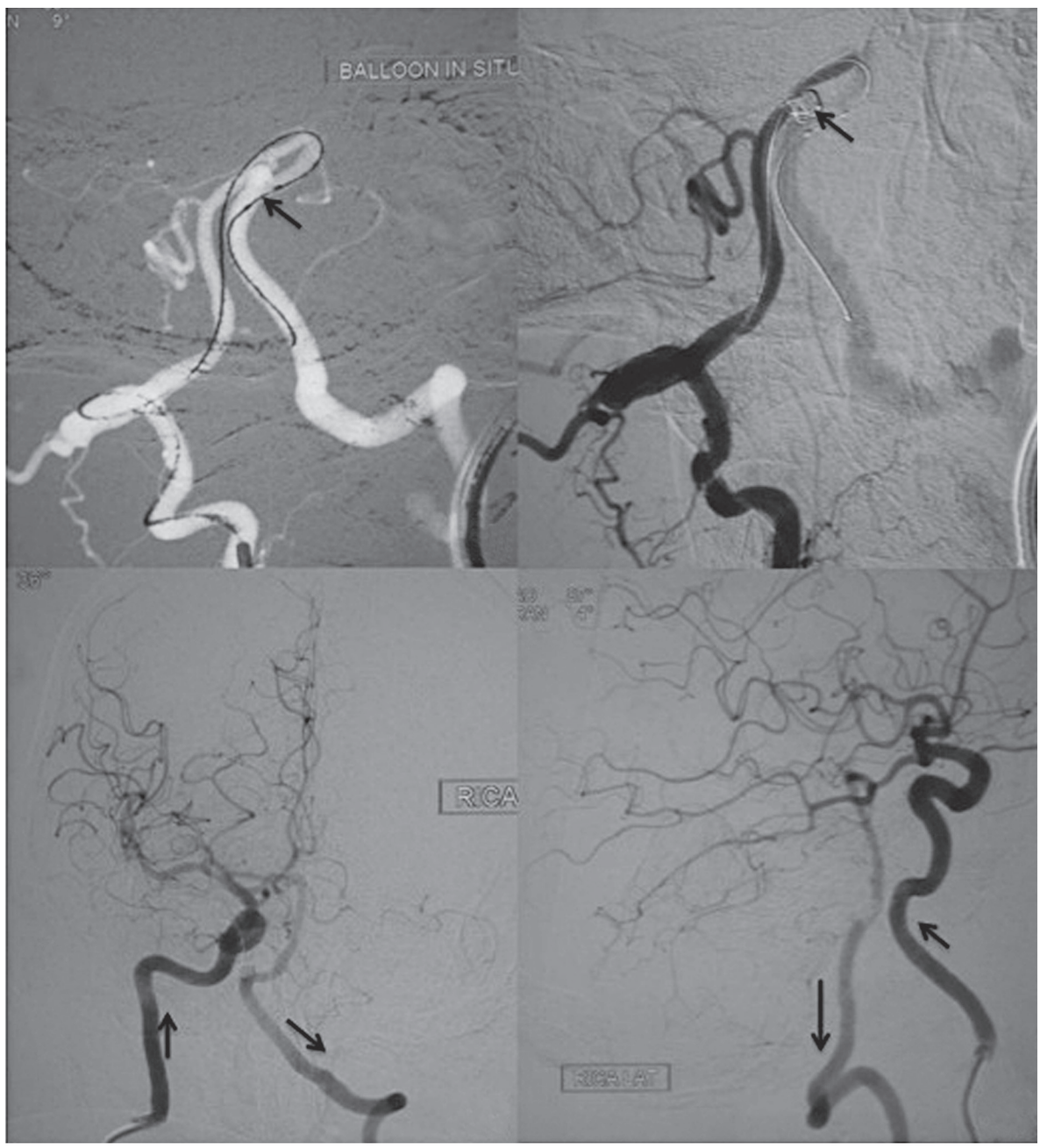

Figure 4 Procedural (A,B) and post procedure (C,D) images of the same patient as in Figure 3. A) Right vertebral angiogram showing a Hyperglide balloon passing from the right vertebral artery into the left vertebral artery (arrow) across the basilar artery distal to the fenestrated segment since the left vertebral artery origin was blocked. B) Image showing complete occlusion of the aneurysm with preservation of both fenestrated limbs (arrow). Right internal carotid artery (ICA) AP (C) and lateral (D) angiograms depicting flow of contrast from the ICA through the posterior communicating artery into the basilar artery and retrograde into the left vertebral artery showing subclavian steal phenomenon (arrows in C and D).

with preservation of both the limbs (Figure 1C,D). No procedure-related complications were encountered and the patient was discharged on the fifth post procedure day in good condition.

Case 2

A 48-year-old man presented with symptoms of acute SAH which was confirmed on brain NCCT (Fisher grade 2). No neurological defi-

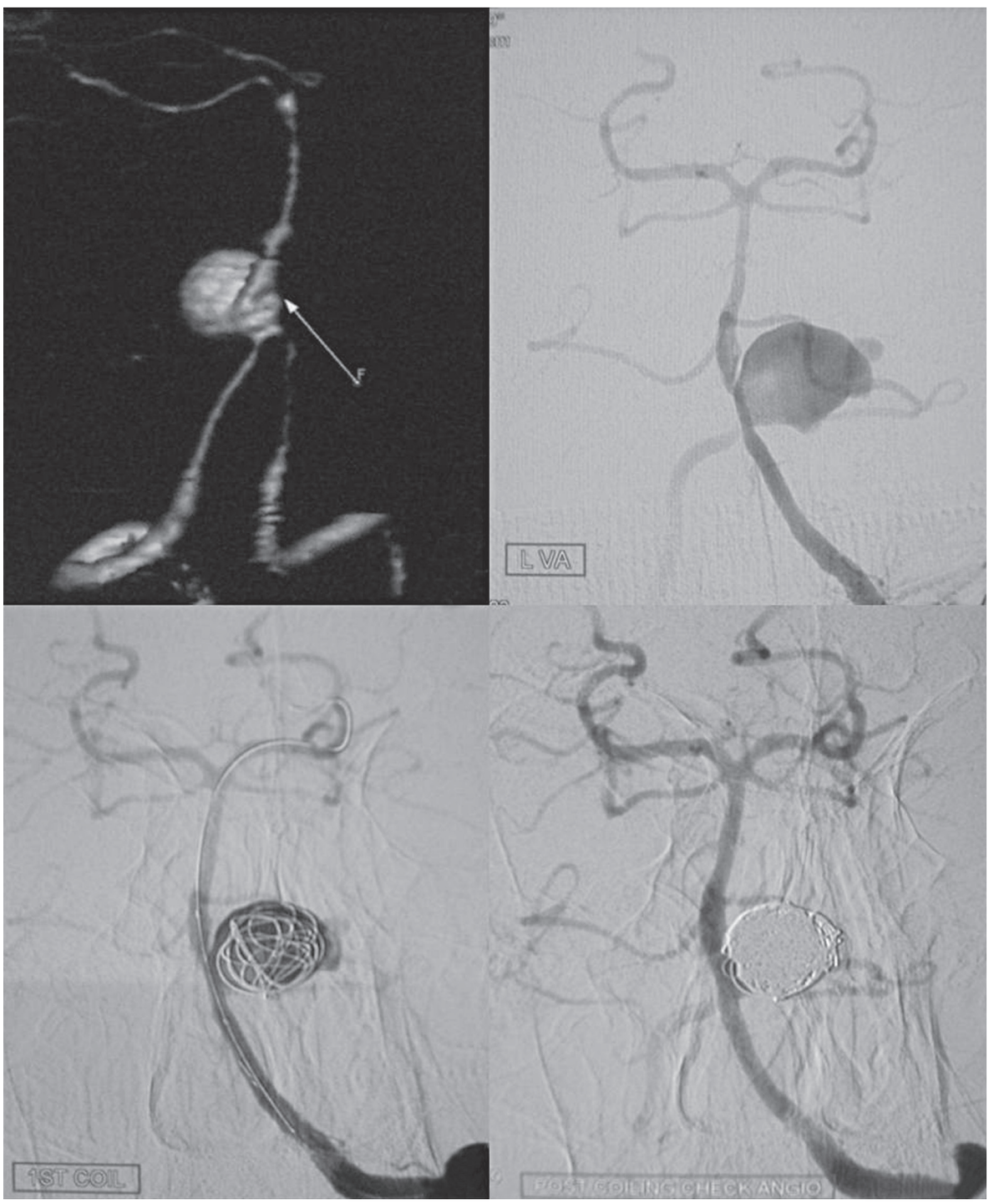

Figure 5 CT angiogram (A) and angiogram (B) showing a large aneurysm arising from the proximal bifurcation of the fenestration (arrow in A). Angiogram during (C) and after (D) coil embolization showing complete obliteration of the aneurysm.

cits were present with a GCS of 15. CTA and cerebral DSA revealed a 10×7×7 mm sized VBJ fenestration aneurysm directed anteriorly apparently arising close to the proximal arterial bifurcation (Figure 2A-C). The aneurysm was successfully embolized using six bare platinum Axium coils (EV3 Neurovascular, Inc, Irvine, CA, USA) with balloon remodeling technique

(Figure 2E). Both limbs remained patent (Figure 2D,F) and the patient was discharged on post procedure day 3 in good general condition. Control DSA at one-year follow-up revealed no recanalization.

Case 3

A 72-year-old man presented with sudden onset headache and drowsiness. On examination, there was meningismus with no associated neurological deficits. NCCT showed Fisher grade 3 SAH (Figure 3A). CTA revealed a 4×4×3 mm sized bilobed aneurysm located at the proximal bifurcation point of VBJ fenestration (Figure 3B). Arch aortogram showed progressive narrowing of the left subclavian artery with abrupt termination at 2.5 cm beyond its origin and proximal to the origin of left vertebral artery. There was a subclavian steal phenomenon whereby the blood was redirected from the right vertebral artery towards the left vertebral artery and into the left subclavian artery side with no flow into the basilar artery and posterior cerebral arteries (PCAs) as demonstrated on right vertebral angiogram. The bilobed aneurysm arose from the proximal bifurcation point of the VBJ incorporating the left limb more and was directed anteriorly (Figure 3C,D). The PCAs and the basilar artery received blood from bilateral internal carotid arteries (ICAs) via dilated posterior communicating arteries (PCAs). The left vertebral artery route was optimal for coiling since the neck was on the left limb of fenestration, but since left vertebral artery origin was occluded the right vertebral arterial route was chosen and a Hyperglide balloon was navigated retrogradely towards the left side across the distal communication point of the VBJ to cover the aneurysm neck (Figure 4A). Embolization was subsequently performed using two Axium coils (EV3 Neurovascular, Inc, Irvine, CA, USA). Control angiogram revealed complete aneurysm occlusion (Figure 4B). The patient developed gait ataxia during the second hospital day. MRI showed a right cerebellar infarct. However, he gradually improved and was discharged with mild residual symptoms after one week.

Case 4

A 34-year-old man presented with symptoms of SAH. NCCT revealed grade 4 SAH. Magnetic resonance imaging examination revealed a large flow void in relation to the left distal vertebral artery impinging on the ipsilateral olive and medullary pyramid. CTA and DSA confirmed the origin of the aneurysm to be from the left limb of a small VBJ (Figure 5A,B). A similar technique of balloon remodeling was employed to embolize the aneurysm using seven coils with preservation of both limbs of the VBJ. Post procedure control DSA revealed a satisfactory result (Figure 5C,D). A six months follow-up DSA was unremarkable.

Discussion

The incidence of fenestration in the presence of a vertebrobasilar junction aneurysm is relatively high – approximately 35% [5]. Thus, when a vertebrobasilar junction aneurysm is present, an associated fenestration should be strongly suspected.

The VBJ fenestration arises as a result of persistence of one of the proximal temporary bridging arteries between embryological bilateral longitudinal neural arteries that aid in the formation of the basilar artery [7]. The lateral walls of the fenestrated artery have a normal intrinsic architecture. The medial walls, however, have focal defects at both ends of the fenestration that make these segments weak and prone to aneurysm formation similar to cerebral artery bifurcations. Here, the media layer is absent and in addition, the sub-endothelium is thin and there is discontinuity of the elastin layer [8]. Another hypothesis for the formation of aneurysms at sites of fenestration is the turbulence of flow at the site of the proximal bifurcation defect of the tunica media [9]. Other proposed explanations have been hemodynamic stresses, genetic [10] and other environmental factors.

Van Rooij et al. [1] classified the simultaneous occurrence of intracranial fenestrations and aneurysms into three types; Type I – aneurysm is located on the proximal aspect of the fenestration, Type II – aneurysm is located on one of the limbs of the fenestration and Type III – aneurysm and fenestration are located on different arteries. According to this classification, all our cases fall into Type I as the proximal aspect of the fenestration was the site of the aneurysm in all cases with or without incorporation of adjacent limbs. This is in agreement with medical literature whereby most authors have reported the proximal aspect of the fenestration to be the common site of aneurysm formation [1]. Interest-

ingly, in all our cases fenestration was detected pre-operatively on CT angiography studies, which were done to look for the cause of subarachnoid hemorrhage. CTA was further complemented by DSA studies including 3D rotational angiography (3DRA) in three patients where exact display of the entire anatomy was possible. Also, morphologically three out of four aneurysms in this series were bilobed, which was also seen in other reported aneurysms at

Table 1 **Comparative data of cases of aneurysms of the vertebrobasilar junction fenestration reported in medical literature.**

Studies	Year of publication	No. of patients	Endovascular procedure	Technical Success/ Occlusion rates	Remarks
Peluso et al.[4]	2007	7	Coiling (balloon assistance, stent, limb occlusion)	100%	4 – good outcome; 2 – died; 1 - dependent
Yoon et al.[13]	2004	4	Coiling	100%	1 vertebral thrombosis (asymptomatic)
Graves et al.[14]	1996	3	Coiling	67%-complete 33%-incomplete	All – good outcome
Albanese et al.[15]	2009	1	Stent-assistance with balloon remodeling through 1 limb; 2nd limb sacrificed	100%	
Nakau et al.[16]	2007	1	Coiling	100%	Aneurysm perforation + parent artery thrombosis; managed appropriately
Ezaki et al.[17]	2003	1	Coiling	100%	
Consoli et al.[18]	2012	2	Stenting (flow diversion)	100%	
Kubo et al.[19]	2005	1	Coiling	100%	Intraventricular tPA given
Picard et al.[22]	1993	2	Detachable latex balloons	100%	1 died due to pulmonary emboli
Pierot[23]	1996	1	Coiling	100%	
Tasker et al.[24]	1997	1	Coiling	NA	
Nakstad et al.[25]	1998	4	Coiling	100%	
Luo et al.[26]	2001	1 (2 aneurysms)	Coiling	100%	
Islak et al.[27]	2002	8 (proximal basilar fenestration)	Coiling (balloon assisted - 5)	87.5% (7/8)	One recanalized aneurysm recoiled after 5 years
Saatci et al.[28]	2002	1	Coiling	100%	
Kai et al.[29]	2006	2	Coiling; 1 limb sacrificed in 1 patient	100%	
Peltier et al.[30]	2006	1	Coiling	100%	
Fujimoto et al.[31]	2007	2	Coiling	(100%)	
Gruber et al.[32]	2010	1	Stent-assisted coiling	100%	'Waffle-cone' technique
Kan et al.[33]	2012	1	Stent-assisted coiling	100%	
Tanaka et al.[34]	2012	4/8	Coiling	100%	
Present study	2012	4	Balloon remodeling	100%	(3/4) 75% - good (1/4) 25% - no follow-up

this location. Surgery had been the conventional mode of treatment of such aneurysms in the pre-endovascular era. However, access to the VBJ is difficult and hampered by the petrous bone. The direct proximity of the aneurysm to the brain stem, perforating arteries and lower cranial nerves makes the approach dangerous. Lateral approaches directed through parts of the petrous bone have been used sometimes for direct access to the VBJ [11,12]. With the advent of the endovascular approach, nearly all such aneurysms command coiling/stenting as the first line of management [13-19]. There are, however, certain restrictions and limitations involved with the endovascular approach as well. It is extremely important to understand the anatomy of the region with a clear depiction of the aneurysm sac, neck and its relation to the fenestration and the parent artery microanatomy [20]. This can be very well appreciated on a 3DRA. As this region is close to vital structures, a methodical approach is warranted.

In all our cases, the neck of the aneurysm was reasonably narrow compared to the dome so that adequate coil positioning was achieved. We used balloon assistance in three patients while in one patient a balloon was kept in the parent artery but its inflation was not required during the procedure. The balloon was positioned in the limb, which had greater incorporation into the aneurysm neck. We were thus successful in preserving both the limbs of each fenestration and at the same time achieving complete aneurysm occlusion. None of the patients required stent placement or limb sacrifice. If possible it is desirable to preserve both limbs of the fenestration segment as both the limbs may give rise to brainstem perforators [13,14]. Also it is important to remember that the size of the limb is not a predictor of the number of perforators it might yield. It is possible that each limb yields some perforators to the ipsilateral halves of the brain stem [21]. One patient (case 3) had subclavian steal phenomenon due to occlusion of left subclavian artery origin, with the left upper limb being perfused by blood from the right vertebral artery and anterior circulation which could be a possible reason for aneurysm formation at this location, in addition, of course, to its location on the weakened wall of the fenestration. A similar procedure was adopted in this patient except that the balloon was positioned on the left side after maneuvering across the vertebrobasilar junction due to the aneurysm neck appearing more lateralized towards the left vertebral arterial end of the fenestration. The aneurysm was approached from the right side of the fenestration and was successfully coiled.

We did an extensive PUBMED review in search of the medical literature regarding the endovascular approach adopted for treating VBJ fenestration aneurysms and found 53 cases in all, described in 21 studies, most of them as reports of single cases [4,13-19,22-34] (Table 1). Thorough evaluation revealed unassisted coiling performed in most of the cases (35 in number), balloon remodeling in about 11 cases and stent usage (as sole or supportive means) in six patients. The combined technical result in all such cases was above 90% with the major causes of morbidity and mortality related to the disease per se i.e. grade of subarachnoid hemorrhage, rather than procedure-related. Regarding the result of direct surgery in this category of patients, a related study [34] has reported a high incidence of cranial nerve palsies in this treatment group, which is conspicuous by its absence in the endovascular group. Most of the institutions worldwide have currently adopted an intra-arterial approach to such lesions unless there is a strong contraindication for the same. The present analysis reveals that the largest series was reported by Islak et al. [27] and Peluso et al. [4], with eight and seven patients respectively. Both these authors used balloon assistance in most patients (in five cases by Islak et al.) with reasonable overall results (11/15 faring well). Our series is currently the third largest to date along with those of Yoon et al. [13], Naksted et al. [25] and Tanaka et al. [34] with four patients each, all showing good technical results. Unlike the other studies, we used balloon remodeling in three patients with no resultant rupture. Stent was not considered by our group due to reservations regarding the strength of the wall of the fenestrated artery and the steep gradient between the parent vessel and the limb of the fenestrated limb. There is presently scant but convincing literature favoring balloon remodeling as the endovascular procedure of choice in appropriately chosen patients of VBJ fenestration. We would like to emphasize that endovascular coiling is a safe and feasible option for VBJ fenestration aneurysms with good technical and clinical outcomes. With good depiction of the anatomical details by contemporary modalities like multi-detector CT angiography and 3D rotational angiography and thorough planning prior to beginning the procedure, the success rates are impressive.

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